

Original Research Article

Role of surgery in the management of mycetoma foot

Beulah Roopavathana S.*, Abinaya R. N., Titus D. K., Inian Samarasam, Cecil T. Thomas, Suchita Chase, Sukria Nayak

Department of General Surgery, Christian Medical College and Hospital, Vellore, Tamil Nadu, India

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***Correspondence:**

Dr. Beulah Roopavathana S.,

E-mail: beulah@cmcvellore.ac.in

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ABSTRACT

Background: Mycetoma is a chronic granulomatous infection of skin, subcutaneous tissue and bone caused by either fungi (eumycetoma) or bacteria (actinomycetoma). Here we present our experience in the management of this rare condition.

Methods: Patients managed for mycetoma foot from August 2011 to January 2018 in authors' general surgery department were included. The data was collected retrospectively from the hospitals electronic database.

Results: Eight patients were recruited with the mean age of the patient was 52 years and had male predominance. Most common clinical presentation was swelling with discharging sinuses in the foot. Imaging performed were X ray foot and MRI. Osteomyelitis was noted in 4 patients (50%). Eumycetoma and actinomycetoma were equal in distribution. Out of 4 patients with eumycetoma, one had grown *Acremonium* species in the culture and out of 4 actinomycetoma patients, 3 had grown *Actinomyces pelletieri* and one had *Nocardia*. Surgical procedures performed were 3 biopsies (37.5%), 3 debridement (37.5%) and 2 amputations (25%). Eumycetoma patients were treated with Itraconazole monotherapy and actinomycetoma patients were treated with Trimethoprim-Sulfamethoxazole and cyclical administration of amikacin. Good response was noted in 6 patients and two patients with Eumycetoma had progression of disease with medical therapy and underwent amputation.

Conclusions: High clinical suspicion and tissue for histopathological and microbiological evaluation is mandatory to confirm the diagnosis. Medical therapy plays a major role in the treatment of mycetoma foot even if the bone is involved. Surgical procedures like debridement and amputation are indicated only for failed medical therapy with progression of disease (seen most often with eumycetoma) and if there are secondary bacterial infections.

Keywords: Actinomycetoma, Eumycetoma, Mycetoma, Welsh regimen

INTRODUCTION

Mycetoma is a chronic suppurative granulomatous infection of skin, subcutaneous tissue and bones caused by filamentous fungi or bacteria.¹⁻⁵ It is endemic in tropical and subtropical countries like Mexico, Africa and India.^{1,2} It is classified into two broad categories, Eumycetoma and Actinomycetoma based on the etiological agent. Eumycetoma is caused by true fungi

and actinomycetoma is caused by higher filamentous bacteria. In India eumycetoma is more common in north India and actinomycetoma is more common in south India.⁶

This condition is seen most commonly in men and in agricultural workers who have a tendency to walk bare feet and exposed to soil. The traumatic inoculation of these organism into the subcutaneous tissue will lead to

this disease process.^{4,7} Host, pathogen and environment plays a major role in the pathogenesis of mycetoma. Following inoculation, there is a nonspecific inflammatory initial response which later becomes more organized and cellular. At an early phase it forms a nodule, abscess or induration which can rupture into fascial planes resulting in deep penetration of disease or it can rupture externally forming sinuses which discharge granules. The affected area will become edematous, hyperpigmented and once the bone is involved it will lead to deformity.⁸ Most commonly foot is involved but actinomycetoma can occur in cervicothoracic region, abdominal wall and perineum. The classic clinical trial of mycetoma is painless soft tissue swelling, draining sinuses and extrusion of grains.⁴

Grains are important as based on the grain colour the causative organism can be diagnosed. Imaging like X ray and MRI is useful to know the involvement of bone and extent of the disease. Deep surgical biopsies under aseptic conditions are needed to obtain tissue and grains for histopathological and microbiological confirmation.⁹ Surgery is an integral component in the diagnosis and treatment of mycetoma. Surgical options include wide incision biopsy, excision of nodule, debridement or amputation. As medical therapy plays a major role, indications of surgery are limited which include biopsy for diagnosis, debridement for extensive infections which has failed medical therapy or with secondary bacterial infections, amputation for secondary bacterial infection with sepsis or extensive bone involvement with failed medical treatment.¹⁰ Various antibiotics are tried for actinomycetoma and combination of Trimethoprim-Sulfamethoxazole and Amikacin (Welsh regimen) has found to be effective with a cure rate of 90% for actinomycetoma. Azoles were found to be effective for eumycetoma and Itraconazole for long duration with surgical excision of residual disease has shown favourable outcome for eumycetoma.¹¹ The failure and recurrence rates are high in patients with Eumycetoma even with long term antifungal treatment combined with surgery.¹² There are no standard guidelines on the duration of medical treatment for mycetoma, so it is important to assess the disease response clinically and radiologically. Response to medical treatment is defined as clinically disappearance of the swelling or nodules, healing of sinuses, skin changes returned to normal, absence of grains in needle aspirates and radiological improvement consists of absorption of the sclerotic bone and reappearance of the normal trabecular pattern as well as bone density.¹³

METHODS

This is a retrospective study conducted in department of general surgery. The study period was from August 2011 to January 2018. Patients who were diagnosed to have mycetoma foot and managed in our general surgery department in the given study period were recruited. 8 patients with mycetoma foot who underwent surgical

procedures like biopsy, debridement and amputation with a confirmed tissue diagnosis of mycetoma only were included. Patients with negative culture or biopsy report for mycetoma or patients with mycetoma at different anatomical location were excluded.

All the 8 patients were followed up for a minimum duration of 9 months to a maximum duration of 7 years. The data was collected from the hospitals electronic database and patient follow up was done from the outpatient charts and telephonic follow up was also performed. Demographic data, clinical presentation, imaging, histopathology and culture report, surgery performed, and medical therapy given with a follow up data for treatment response were tabulated and analyzed. The treatment response was based on clinical examination and radiological findings. The data analyzed were presence of osteomyelitis, etiological agent of mycetoma, medical therapy which includes antibiotic and antifungal based on the etiological agent, duration of medical therapy, types of surgical procedure performed and its indication.

RESULTS

Eight patients with mycetoma foot in the given period whose diagnoses were confirmed with histopathological and microbiological examination were recruited. Out of 8 patients, 7 were male (87.5%) and one was female (12.5%). The mean age group of patients in the study group was 52 years. 50% of the patients were farmers by occupation and two patients had history of thorn prick in the bare foot preceding the infection. Two patients were diabetic, and one patient was on immunosuppressants following renal transplant. All the 8 patients had a clinical presentation of foot swelling with discharging sinuses as shown in Figure 1. Two patients had grains noted in the discharge. The duration of symptoms varied from 2 years to 5 years. 2 patients had biopsy done at another centre and started on antifungal for mycetoma foot prior coming to authors' centre. Patient demographic details and disease specific characteristic features are described in Table 1.



Figure 1: Left foot swelling with multiple discharging sinuses in a patient with mycetoma foot. (A): involvement of plantar and medial aspect, (B) involvement of dorsal aspect.

Table 1: Patient demographics and disease specific characteristic features.

Patient characteristics	Number n (total-8)
Gender	
Male	7
Female	1
Occupation- agriculture workers	4
Comorbidities	
Diabetes mellitus	2
Post renal transplant	1
Swelling with discharging sinuses	8
Osteomyelitis	4
Etiological agent	
Eumycetoma	4
Actinomycetoma	4

Imaging was performed for all patients, X ray foot for 4 patients and MRI for 4 patients. Osteomyelitis was noted in 4 patients (50%). Surgical procedures performed were 3 biopsies (37.5%), 3 debridement (37.5%) and 2 amputations (25%). Two patients with osteomyelitis showed good response with medical therapy as shown in Figure 3. One patient with osteomyelitis on medical therapy, Itraconazole had worsening (Figure 2) and underwent amputation. One post renal transplant patient who had amputation of first and second toe in the past was diagnosed to have mycetoma foot at another centre and given medical therapy, Itraconazole for a year. He presented with osteomyelitis (Figure 4) and secondary bacterial infection with failed medical treatment and he underwent below knee amputation.



Figure 2: Pre and post medical therapy imaging in a patient with mycetoma foot, (A): MRI foot showing soft tissue and bone involvement (base of metatarsal and tarsal) pre medical treatment, (B): X ray foot post medical treatment with worsening, underwent amputation.

Eumycetoma and actinomycetoma were equal in distribution (4 each). Out of 4 patients with eumycetoma, one had grown *Acremonium* species in the culture and out of 4 actinomycetoma patients, 3 had grown *Actinomyces pelletieri* and one had *Nocardia*. Eumycetoma patients were treated with itraconazole

monotherapy for a minimum duration of one year and maximum up to 3years, except one patient who had grown *Acremonium* species received voriconazole for 8weeks followed by itraconazole. Actinomycetoma patients were treated with trimethoprim-sulfamethoxazole for a duration of 6months to one year and 3 cyclical administration of Amikacin based on Welsh regimen. Good response was noted in 6 patients and two patients with Eumycetoma had progression of disease with medical therapy as mentioned earlier underwent amputation.



Figure 3: X ray images of a patient with mycetoma foot before and during medical therapy. (A): X ray foot oblique view showing metatarsal involvement before treatment, (B): X ray foot oblique view showing stable disease with mild response during medical therapy for 6 months duration.

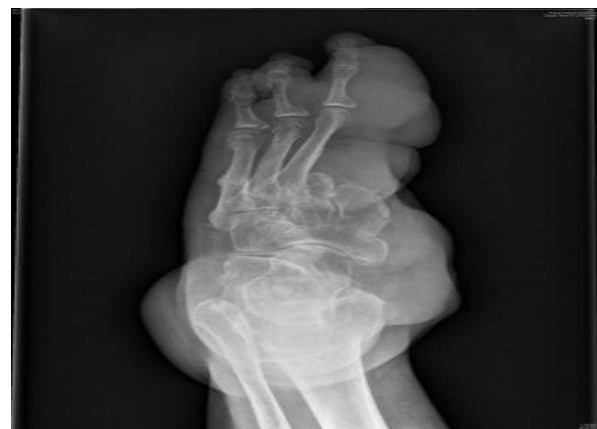


Figure 4: X ray foot of a post renal transplant patient who had amputation of first and second toe in the past and now presented with osteomyelitis and secondary bacterial infection following failed medical therapy with Itraconazole for one year.

DISCUSSION

Mycetoma is a chronic suppurative granulomatous infection of skin, subcutaneous tissue and bones caused by traumatic inoculation of either filamentous fungi or bacteria.^{2,4,14} It was first described in Madurai in South India.⁷ India is an endemic area for this neglected tropical disease. Due to indolent course and rarity of this condition there is always a diagnostic delay. In present

study group, we had male predominance and it was most commonly seen in farmers as described in the literature.^{4,7}

Eumycetoma and actinomycetoma had equal distribution and all patients presented with swelling of foot and discharging sinuses which should raise high index of clinical suspicion. Bone involvement was noted in 50% of the patients as described due to the natural pathogenesis of the disease which can spread deeply into fascial planes and can involve bone.⁸ If not diagnosed early, it can lead to permanent deformity due to extensive bone involvement. All studied patients had diagnosis confirmed with histopathological and microbiological examination of tissues either following biopsy or debridement. As mentioned in literature deep tissue biopsy with grains are mandatory to confirm the diagnosis. Medical therapy was the first line of treatment in all 8 patients in present study group which is comparable with the literature.^{4,11} Standard medical therapy like Itraconazole for Eumycetoma and Welsh regimen for actinomycetoma showed a good response except in two patients with Eumycetoma who had failed medical treatment with disease progression and secondary bacterial infections. So, surgery is performed only for selected indications as mentioned in the literature. In present study group, surgical procedures like open incision biopsy and limited debridement were performed for tissue diagnosis and amputation was performed for failed medical therapy with progression of disease in patients with eumycetoma.

CONCLUSION

Mycetoma foot is a rare chronic infection which can lead to permanent deformity and needs high index of clinical suspicion for early diagnosis. Diagnosis is confirmed by histopathological and microbiological examination. Medical therapy is the standard protocol and surgery is indicated only in few complications like secondary bacterial infections and failed medical therapy with progression of disease (most often seen with eumycetoma). Osteomyelitis is not a contraindication for medical therapy. Radical surgery like amputation can be avoided if diagnosed early.

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Ethical approval: The study was approved by the Institutional Ethics Committee

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